

[CASE REPORT]

The Exacerbation of Hemicrania Continua Mimics Trigeminal Neuralgia

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Abstract:

We report the case of a 46-year-old man with hemicrania continua presenting as exacerbations mimicking trigeminal neuralgia. The patient was tentatively diagnosed with trigeminal neuralgia, and treatment with various combinations of drugs was performed after the onset of pain. However, when the condition of the patient did not improve, we suspected hemicrania continua, and treatment with indomethacin was initiated. There was a marked alleviation of his pain within 24 hours. Thus, clinicians should be aware that the duration and frequency of exacerbations of hemicrania continua are variable.

Key words: trigeminal autonomic cephalalgias, indomethacin, tooth extraction

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Introduction

According to the third edition of the International Classification of Headache Disorders (ICHD-3), hemicrania continua (HC) is a persistent, strictly unilateral headache, with superimposed exacerbations of moderate or greater intensity that are accompanied by ipsilateral autonomic signs (1). It is often misdiagnosed, even by neurologists, as a migraine, cluster headache, sinus headache, or dental pain because of the great heterogeneity of its pain characteristics (2). Delays in the diagnosis of HC not only reduce patient quality of life but also can lead to unnecessary therapeutic interventions, such as dental extraction, sinus surgery and nerve block injections. In this report, we present a case of HC that occurred after wisdom tooth extraction and show exacerbations mimicking trigeminal neuralgia.

Case Report

A 46-year-old man underwent right upper wisdom tooth extraction due to dental caries. Before extraction, the patient was free from headaches and facial pain. Two weeks later, he developed daily headaches and facial pain, which had the characteristics of continuous pain and two types of superim-

posed exacerbations (Fig. 1). The continuous pain was of moderate intensity and was localized to the right side in the orbital, supraorbital, maxillary, mandibular, and parieto-temporal areas (Fig. 2A) (3). The pain was characterized as stabbing or throbbing, and was aggravated by routine physical activity and alcohol consumption. The first type of superimposed exacerbation was more severe pain that was long-lasting. It was located in the same areas as the continuous pain and occurred every morning and midnight, lasted for 1-2 hours, and was accompanied by right-sided nasal congestion and occasional facial sweating. There were no other autonomic symptoms, such as conjunctival injection, lacrimation, eyelid edema, miosis, and ptosis. Drinking alcohol made the pain worse and the patient became agitated and hit himself with his fist. The second type of superimposed exacerbation was a sudden and lancinating pain that lasted for a few seconds. This pain was located in the right cheek and mandible (Fig. 2B) (3). It occurred over 20 times a day, and its frequency seemed to increase during bouts of the first type of exacerbation. There were no autonomic symptoms during the short-lasting pain. Due to intolerable pain, the patient visited four medical institutions after treatment failure with the over-the-counter drug loxoprofen (Table). The patient was tentatively diagnosed with trigeminal neuralgia (TN), and treatment with various combinations of

drugs, including loxoprofen, famciclovir, diclofenac, pregabalin, carbamazepine, tramadol, prednisolone, intramuscular pentazocine and infraorbital nerve block were tried for two weeks after the onset of pain. However, his symptoms

did not improve. The patient was subsequently admitted to our hospital for the further evaluation of his condition.

The results of a neurological examination were normal. MR imaging, including 3D-T2-weighted driven equilibrium imaging, did not reveal any neurovascular contact between the trigeminal nerve and blood vessels. Upon reviewing the patient's history, we considered the possibility of HC, and indomethacin was initiated at a dose of 25 mg, twice a day. There was a marked alleviation of his pain within 24 hours. The patient had not experienced such improvement with any drug in the past. The dose was titrated to 50 mg, twice a day, over three days, and the patient showed a complete response. Over the next 3 months of follow-up, tapering of indomethacin led to a relapse of both types of reported pain.

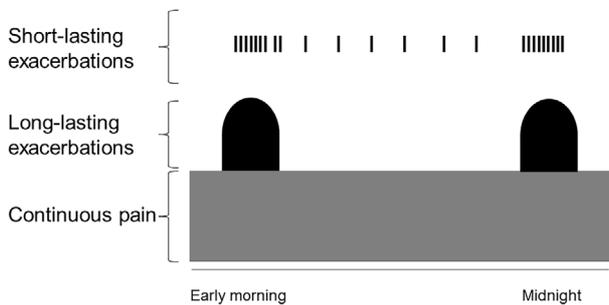


Figure 1. A diagrammatic representation of the pain characteristics in this case. Long-lasting exacerbations, which involved continuous pain that was more severe than the reported continuous pain, occurred every morning and in the middle of the night, and lasted for 1-2 hours. The short-lasting exacerbations involved sudden and lancinating pain that lasted for a few seconds. The frequency seemed to increase during the long-lasting exacerbations.

Discussion

The patient noted two types of exacerbations: short-lasting (a few seconds), and long-lasting (1-2 hours). Although the long-lasting exacerbations might be saw-tooth patterns of short-lasting exacerbations, they showed different characteristics. Exacerbations in HC are highly variable in terms of character, intensity, duration, and frequency (4). They can

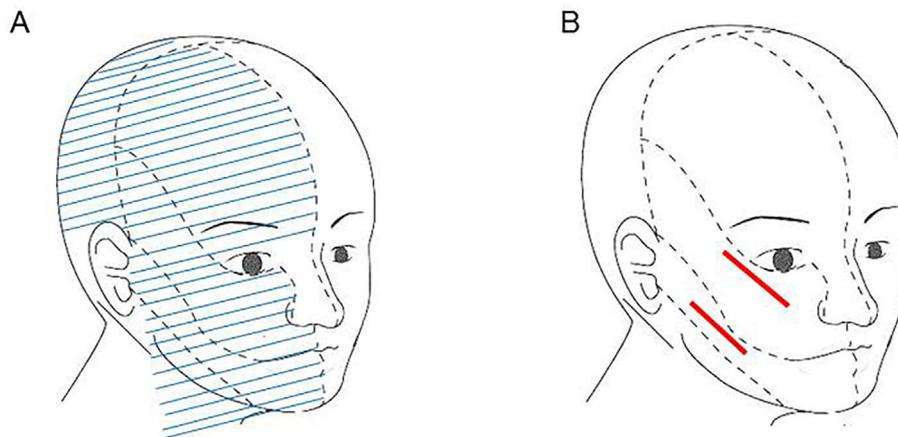


Figure 2. The areas of pain. A: The blue hatched area shows the areas of continuous pain and long-lasting exacerbations. B: The red line shows the areas of short-lasting exacerbations. Modified with permission from ref. 3).

Table. Clinical Course before Admission to Our Hospital.

Day	Type of medical institution	Medication	Surgical treatment
1-	(Over-the-counter drug)	loxoprofen	
4	Dermatologist, Clinic 1	famciclovir, diclofenac	
6	Pain Clinic 2	pregabalin	nerve block
7	Pain Clinic 3	pregabalin, tramadol	nerve block
8	Pain Clinic 3	pregabalin, tramadol	nerve block
10	Neurologist, Clinic 4	pentazocine, carbamazepine, diclofenac, prednisolone	
13	Neurologist, Clinic 4	pentazocine, tramadol diclofenac, prednisolone	
14	Our hospital		

The patient visited 4 medical institutions one after another before admission to our hospital.

mimic almost all primary headaches and/or neuralgias. The patient had autonomic symptoms and agitation during the exacerbations of the pain. Alcohol is a known as trigger of the exacerbation of pain (4). Carbamazepine was ineffective, but indomethacin relieved the pain of our patient. The patient fulfilled the International Headache Society criteria for HC (ICHD-3 code 3.4) (1).

It has been reported that the average delay in the diagnosis for HC is 95 ± 75 months (2, 5, 6). Various reasons can underlie this delay. In our case, the patient was so frightened by the exacerbations that he rarely acknowledged the continuous pain he experienced; thus, attending physicians may not have fully investigated his symptomology. If attending physicians note only short-lasting pain, they might misdiagnose these symptoms as TN. There are only a few reports of neuralgia from the exacerbation of HC (5, 6). Thus, an understanding of both continuous pain and the exacerbations is crucial for reducing the misdiagnosis of HC.

It is difficult to provide clear evidence of a causal relationship between HC and tooth extraction; however, a similar case of cluster headache after molar extraction has been reported (7). The upper wisdom teeth are innervated by unmyelinated C fibers (8). These C fibers project to the trigeminal subnucleus caudalis in the brain stem (9). Experimental tooth pulp denervation is known to result in hyperactivity in expanded receptive fields of neurons in the trigeminal nuclei (9). The reorganization induced by the irritation or deafferentation of C fibers may contribute to the activation of the trigeminal-autonomic reflex, the final common pathway for all TACs. In our case, a latency of two weeks after dental extraction was observed before the onset of HC, which is similar to the latencies observed in previous cluster headache cases (7).

Patients can experience more than one type of headache. The location of multiple headaches and facial pain could

overlap, and attacks can occur simultaneously. Clinicians should be aware that the duration and frequency of exacerbations of HC are variable. Indomethacin should be considered for individuals with a chronic continuous unilateral headache suggestive of HC.

The authors state that they have no Conflict of Interest (COI).

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